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Brown Tumor of the Maxilla in Patient with Primary Hyperparathyroidism

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Abstract | **Clinical Case**

Brown tumors are osteolytic lesions that rarely indicate hyperparathyroidism. They usually occur in the terminal stage of primary or secondary hyperparathyroidism. Over the past three decades, the diagnosis of hyperparathyroidism has often been made during the asymptomatic phase, thanks to systematic calcium and parathyroid hormone levels. We report the case of a patient with primary hyperparathyroidism revealed by a maxillary tumor, whose CT scan revealed an aggressive osteolytic process. The anatomopathological examination revealed a benign giant cell tumor of the maxilla. The diagnosis of a brown tumor was suggested and confirmed after a phosphocalcic workup indicated hypercalcemia and hypophosphatemia. The etiological investigation revealed a right retro-thyroid nodule on cervical ultrasound, which after biopsy excision of the right inferior parathyroid gland confirmed the presence of a right inferior parathyroid adenoma, associated with homolateral lobectomy. A parathyroid hormone level of 1178 pmol/L (18 times the normal level) confirmed the diagnosis. This case study highlights the challenges in accurately diagnosing patients with osteolytic processes in the maxilla and underscores the necessity of investigating hyperparathyroidism in the presence of a giant cell lesion, given the insidious nature of this condition.

Keywords: Brown Tumor, Hyperparathyroidism, Giant Cell Tumor, Maxilla, Parathyroid Adenoma.

INTRODUCTION

The brown tumor is a rare pathology seen by maxillofacial surgeons resulting from a disturbance in bone metabolism associated with primary or secondary hyperparathyroidism.

Its origin is directly linked to the action of parathyroid hormone on bone. In most cases, it is secondary to primary hyperparathyroidism.

It is diagnosed in approximately 4.5% of patients with primary hyperparathyroidism, and in 1.5-1.7% of those with secondary hyperparathyroidism [3].

Albright first described this tumor in the facial skeleton in 1934 [5]. Although it can affect the entire skeleton, its involvement in the maxillo- mandibular region remains uncommon, manifesting as a progressive, painful jugal swelling. It can be locally aggressive, but has no metastatic potential

Histologically, it belongs to the giant cell lesions of the maxilla.

Therefore, diagnosis of certainty requires systemic investigations: calcium and parathyroid hormone (PTH) assays to differentiate between these lesions.

Primary hyperparathyroidism is characterized by an overproduction of PTH, mainly due to adenomas in 85% of cases [1]. It is generally detected by elevated calcium and decreased phosphate levels in routine blood tests.

Less than 5% of cases are diagnosed by the presence of brown tumors [4]. Clinical symptoms include kidney pain caused by kidney stones, as well as gastrointestinal and neurological intestinal and neurological disorders [11].

Secondary hyperparathyroidism results from chronic renal failure.

Most patients with a history of dialysis or renal failure are suspected of suffering from it. In such cases, patients present with hypocalcemia and hyperphosphatemia, as opposed to the values observed in primary hyperparathyroidism.

In both types of hyperparathyroidism, levels of the enzyme alkaline phosphatase remain normal, except in cases of advanced osteolysis, while circulating circulating PTH levels are elevated.

Patients with secondary hyperparathyroidism do not show symptoms of hypercalcemia, but may present signs associated with overproduction of PTH, such as brown tumors and fibrocystic osteitis, also known as renal osteodystrophy.

CLINICAL CASE

A 56-year-old patient consulted our department for a painful right buccal swelling that had been evolving for 1 month. The examination revealed no particular pathological history.

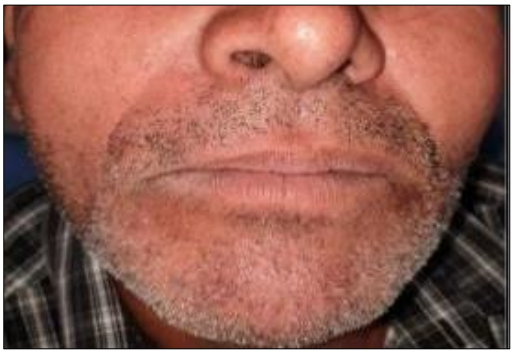

Fig. 1: Extra-oral examination

Intra-oral examination revealed (figure 2): Insufficient oral hygiene a right gingiva-maxillary tumefaction filling the right upper vestibule, causing an arching of the palate. The teeth opposite the lesion were alive and non-mobile (positive cold test on 13, 14, 15 and 16).

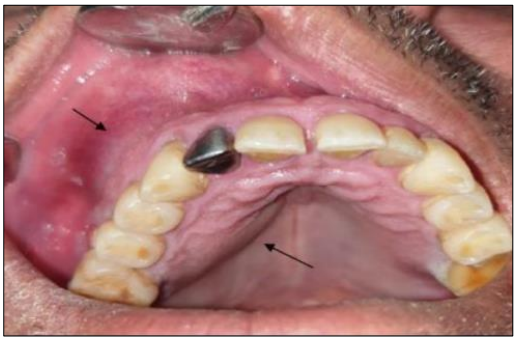

Fig. 2: Intra-oral examination

Panoramic radiograph (figure 3) showed: osteolytic image, partially limited by a border of osteocondensation in the right maxillary premolo molar region.

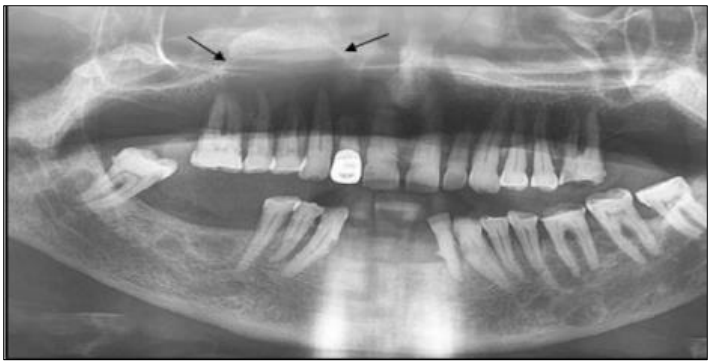

Fig. 3: Panoramic radiograph

Retroalveolar X-ray (figure 4) showed: thinning of the roots of 15 and 14, which were enclosed by the osteolytic lesion, with the beginning of root resorption, particularly at the apices.

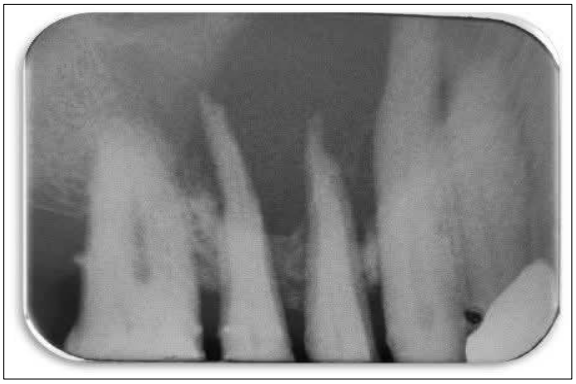

Fig. 4: Retroalveolar X-ray

A CT scan of the facial bones and soft tissues with injection of contrast medium revealed a well-limited osteolytic image of tissue density, enhanced after injection, in the right maxillary premolomolar region, with destruction of the vestibular and palatal bone cortices and limited invasion of the nasal and sinus cavities on the same side (Figure 5).

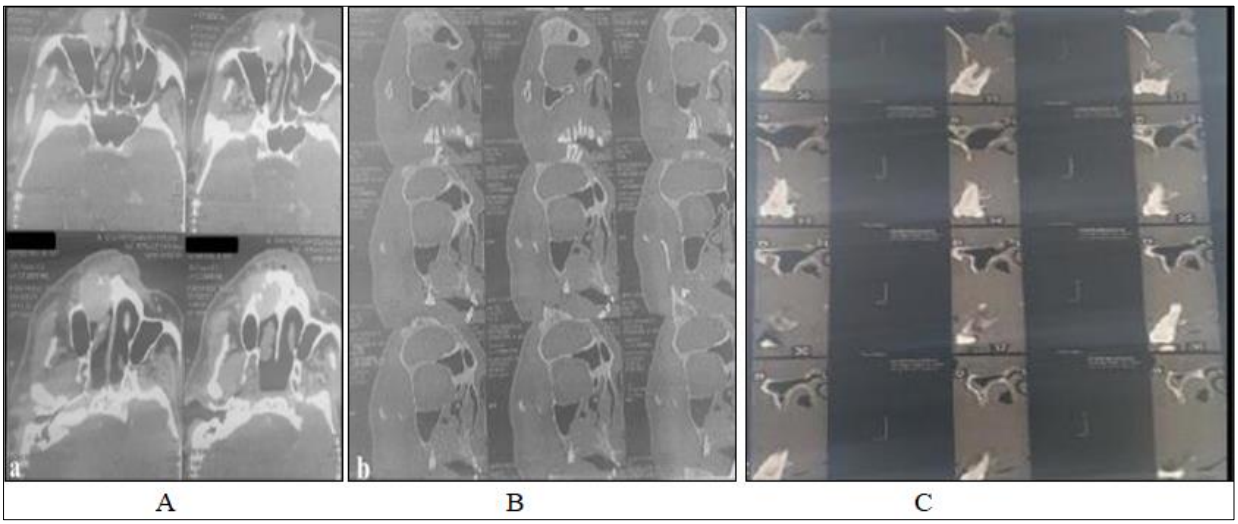

Fig. 5: (A): CT cross-section soft tissue window (B): CT frontal section bone window (C): oblique coronal reconstructions

Based on the clinical and radiological data the following diagnoses were evoked: - True giant cell tumor - Central giant cell granuloma - Brown tumor - Non-ossifying fibroma – Myxoma.

Phosphocalcic workup revealed: Hypercalcemia at 3.4 mmol/l, Hypophosphatemia at 0.45 mmol/l, Alkaline phosphatase at 576 mmol/l, PTH level at 1178 mmol/l (18 x normal 18 times the normal level), decreased vitamin D value at 19. 45 mmol/l.

Cervical ultrasound showed the presence of a right retro- thyroid nodule.

Based on the results of further investigations, a diagnosis of maxillary brown tumor due to primary hyperparathyroidism was made. The patient was referred to the Department of Otolaryngology for management of hyperparathyroidism.

Parathyroid gland scintigraphy revealed a pathological parathyroid with multiple osteolytic foci.

This patient was admitted to the ENT (ear, nose thyroid) department, where he underwent two biopsies under general anesthetic: an excisional biopsy of the right lower parathyroid gland, which revealed the presence of a right lower parathyroid adenoma, combined with a homolateral lobectomy. A biopsy of the maxillary lesion confirmed the diagnosis of a brown tumor.

Biological workup data concerning elevated blood calcium, decreased phosphoremia and increased PTH, in addition to the presence of a parathyroid adenoma, guided us towards the diagnosis of primary hyperparathyroidism. Clinical, radiological and pathological findings confirmed the diagnosis of brown tumor. The final diagnosis of this case was: Maxillary brown tumor due to primary hyperparathyroidism.

Medical treatment consisted of a 4 mg infusion of Zoledronic acid and hormonal correction to reduce

excessive bone resorption.

A check-up was carried out after 2 months and showed an improvement in the phosphocalcic balance: Ca++ : 2.02 mmol/l Phosphorus : 0.7 mmol/l PTH (parathormone) : 45 mmol/l PAL (alkaline phosphatase) : 322 mmol/l.

The patient was put on calperos and sterogyl to promote bone healing and maintain a healthy bone metabolism.

The brown tumor began to regress progressively after regular check-ups. Clinical and radiological follow-up after 6 months showed clear regression of swelling and bone resorption (figures 6 and 7).

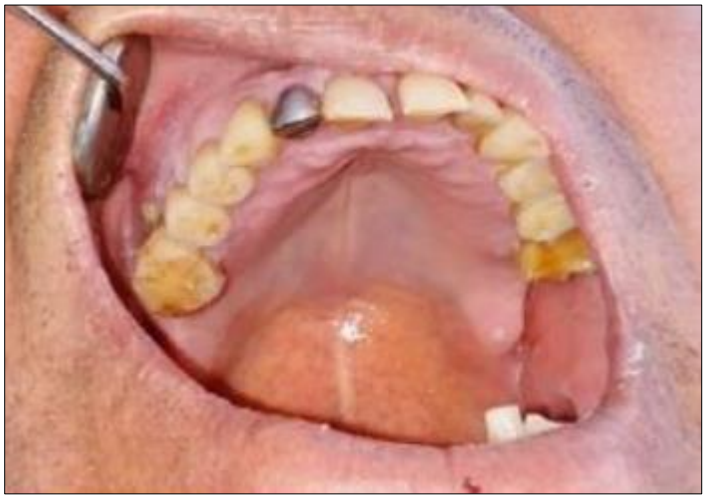

Fig. 6: endo buccal check-up examination

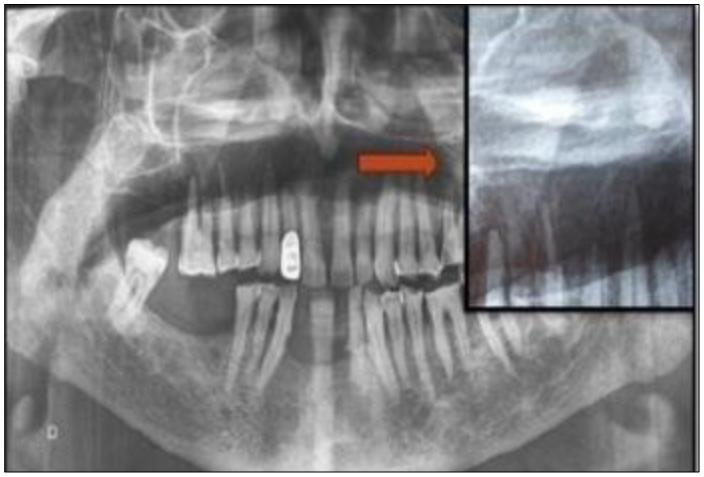

Fig. 7: Right gingiva maxillary tumor filling the vestibule and palate and causing tooth loss and displacement.

DISCUSSION

Currently, hyperparathyroidism is discovered by chance in 75-80% of cases, when a blood test reveals asymptomatic hypercalcemia [10].

Bone manifestations of hyperparathyroidism: bone cysts, osteoporosis, subperiosteal resorption and brown tumors, represent the late expression of the disease, and have become rare, occurring in 5 to 15% of cases [2]. Moreover, it is exceptional for a brown tumor to be the first and only sign of parathyroid hyperfunction.. The incidence of these lesions reported around twenty years ago was 1.5% to 1.7% in secondary hyperparathyroidism and 3% in primary hyperparathyroidism [6]. In the majority of cases, hyperparathyroidism is associated with primary hyperparathyroidism. In over 80% of cases, primary hyperparathyroidism results from a parathyroid adenoma, and less frequently from hyperplasia (15%) (8). Primary hyperparathyroidism often affects patients over the age of 50, particularly postmenopausal women. There is a predilection for females in benign hyperparathyroidism Brown tumors can affect the entire skeleton, with the most frequent localizations being the pelvis, ribs, femurs, mandible and hands. Maxillary localization is extremely rare.

From a clinical point of view, the symptoms induced by these lesions vary according to their size and

location. Indeed, the brown tumor may present with manifestations similar to those of other maxillary tumors and pseudotumors, such as bone expansion in the cheek, palate and/or gingiva, leading to facial deformity and asymmetry, pain, tooth mobility and even tooth loss. In some patients, the lesion may be asymptomatic, and the diagnosis is made incidentally following a routine radiological examination (figure 8).

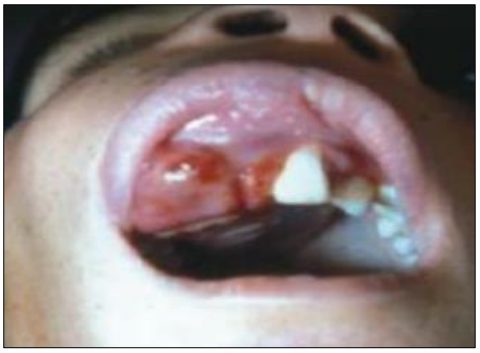

Fig. 8: Right gingiva maxillary tumor filling the vestibule and palate and causing tooth loss and displacement [13].

On imaging, brown tumors appear as clearly delineated osteolytic lesions, presenting dense, heterogeneous areas within the altered bone structure, with notable bone hypertrophy, especially notable in the mandible when associated with fibrocystic osteitis. The CT scan reveals a contrast-enhancing mass of tissue density, with no soft- tissue invasion or periosteal reaction. Often, the lesion fills the maxillary sinus, forming an intrasinus mass (figure 9).

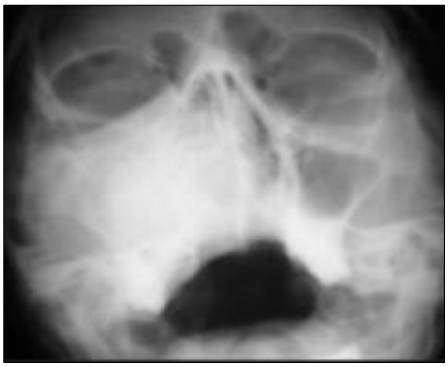

Fig. 9: Blondeau incision, filling of the right maxillary sinus in relation to a gingiva-maxillary tumor [7].

Standard skeletal X-rays are taken to check for other possible localizations, renal lithiasis or nephrocalcinosis. Other radiological features of hyperparathyroidism include subperiosteal resorption, mainly localized to the phalanges. Ultrasound and cervical scans are prescribed to detect any lesions of the parathyroid glands causing hyperparathyroidism (figure 10).

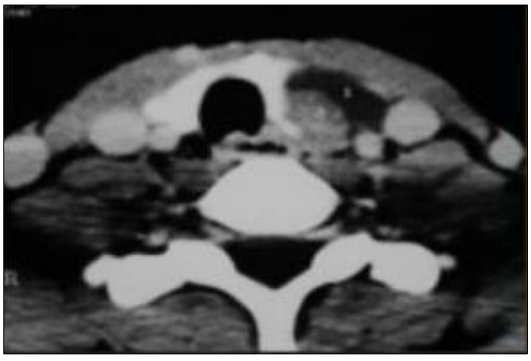

Fig. 10: Axial section cervical CT scan, hypodense left retrothyroid nodule [7].

A CT scan with technetium (Tc-99m) injection is the best way to locate the lesion in the parathyroid glands or ectopic tissues prior to surgery.

The histological characteristics of brown tumors are very similar to those of other giant cell

tumors. The tumor tissue is composed of fibroblastic spindle cells and multinucleated osteoclastic giant cells irregularly distributed around a network of interstitial capillaries.

However, histology alone is not sufficient to confirm the diagnosis of brown tumor, which often requires a combined assessment of the clinical context histological features in favor of giant cell tumors, and laboratory analysis revealing hyperparathyroidism (figure 11).

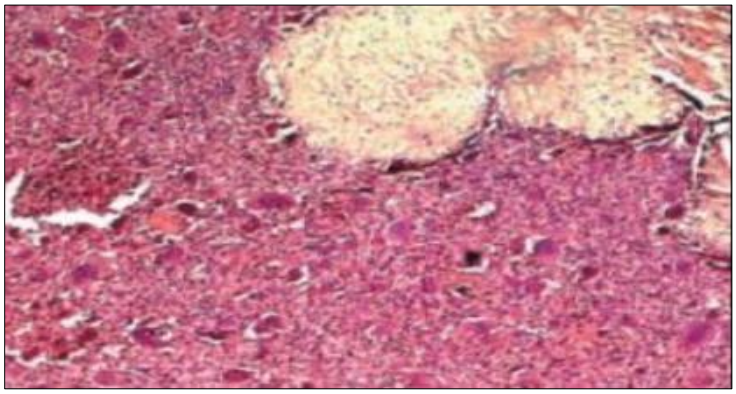

Fig. 11: (HE*40) gingival mucosa with numerous osteoclastic giant cells in the chorion [4]

Treatment of the brown tumor focuses primarily on resolving its underlying cause: hyperparathyroidism. According to most experts, a progressive and spontaneous shrinkage of the tumor is observed after correction of hyperparathyroidism, followed by bone repair with sclerosis and calcifications substituting the original tumor. Indeed, a general consensus holds that parathyroidectomy is the treatment of choice for primary hyperparathyroidism. However, there is a divergence of opinion as to the treatment of secondary bone lesions. In fact, the evolution of brown tumors after parathyroidectomy depends on their composition. Nevertheless, despite PTH treatment, some cases show partial regression or reappearance of the tumor. In general, the treatment of brown tumors is conservative, except in very advanced cases that cause significant aesthetic alterations or are resistant to medical treatment.

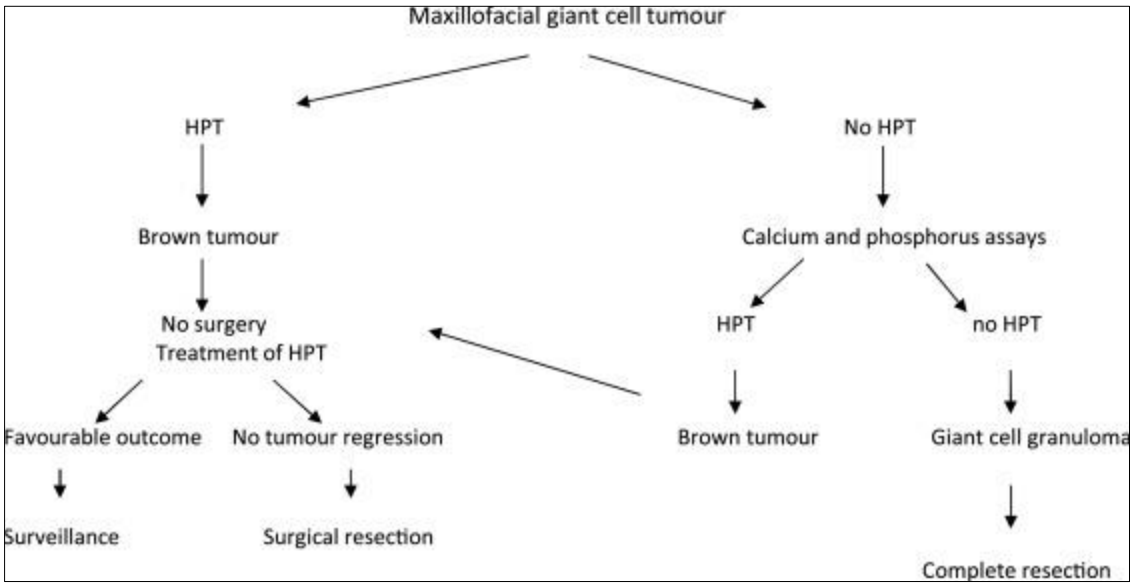

Fig. 12: Management of maxillofacial giant cell tumour [12]

CONCLUSION

Primary or secondary hyperparathyroidism may be manifested 99% of the time by the presence of osteolytic lesions of the facial skeleton. Consequently, a giant cell lesion of the maxilla must be systematically investigated for hyperparathyroidism by means of a phosphocalcic test and a parathyroid hormone assay.

Diagnosis of primary hyperparathyroidism avoids the need to operate on maxillary brown tumors, which should regress after removal of the parathyroid lesion. The present case demonstrated the difficulty of establishing a correct diagnosis in patients with an osteolytic process of the maxilla presenting histologically as a giant cell lesion, which is crucial for

proposing appropriate treatment.

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